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Case Report: Feasibility of Video-Assisted Thoracoscopic Thymectomy in Giant Thymoma

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BACKGROUND: Thymomas are slow-growing mediastinal masses, usually resectable through minimally invasive approaches. However, some thymomas, referred to as 'giant thymomas', can exceed 10 cm and are thought to require open resection. There are few reports of successful removal via video assisted thoracoscopic surgery (VATS) in adults. We present the first case of a giant thymoma resected via VATS in the pediatric population.

CASE PRESENTATION: A 17-year-old female presented with a bilobed, anterior mediastinal mass. Three family members had history of thymoma resected via median sternotomy. Adult thoracic surgery and pediatric surgery collaborated on a bilateral VATS approach with left mini thoracotomy for mass extraction. On gross pathology, the specimen measured 16 cm in maximum dimension and the margins were negative. Epidural analgesia was utilized with minimal postoperative opioid needs. The patient was discharged to home on post-operative day 3.

CONCLUSION: Bilateral VATS with mini-thoracotomy for extraction is a feasible approach for the resection of a pediatric, giant thymoma. Comprehensive pain management, including a thoracic epidural, allowed for excellent pain control and facilitated rapid discharge. Collaboration between adult thoracic surgery and pediatric general surgery was fundamental to the success of this approach.

Keywords: *Thymoma, Thymectomy, Giant Thymoma, Pediatric Surgery, VATS, Video-Assisted Thoracoscopic Surgery, Video-Assisted Thoracoscopic Thymectomy.*

DISCLAIMER

Information published in this article is not intended to replace clinical judgement or establish a protocol for the care of all children with giant thymoma. The treatment described in this article is not intended to be the sole source of guidance in the management of pediatric giant thymoma, and may not provide the only acceptable approach to the management of this condition. The views expressed in this article are solely those of the authors and do not reflect the views of The Pacific Northwest Journal of Surgery or its Editorial Board.

BACKGROUND

Thymoma is an exceedingly rare tumor in the pediatric population, with an incidence of 0.13-0.32 per 100,000 children.[1] Though often sporadic, familial inheritance has been observed in association with a constitutional translocation t(14;20)(q24.1;p12.3).[2] Thymomas usually present as slow growing masses that can invade surrounding structures including the pericardium, trachea, and great vessels, but rarely metastasize.[3-5] Due to the indolent nature of these tumors, patients can be asymptomatic at presentation with an incidental finding on chest radiograph prompting workup.[6] As the tumor grows,

however, it can exert a mass effect, causing respiratory symptoms including shortness of breath, chest pain, wheezing, or cough.[5] This indolent presentation with persistent slow growth can lead to 'giant thymomas', tumors in excess of 10 cm; in fact, thymomas as large as 36 cm have been reported in the pediatric population.[7] Thymomas may also exhibit paraneoplastic syndromes, such as myasthenia gravis, pure red blood cell aplasia, acquired hypogammaglobulinemia, or a connective tissue disorder.[8,9] Work-up for thymoma includes cross-sectional imaging with CT or MRI, though the most common initial imaging is chest radiograph.[10] Laboratory testing should be used to rule out other anterior mediastinal masses, including beta-human chorionic gonadotropin (b-HCG) and alpha-feta protein (AFP) for germ cell tumor and lactate dehydrogenase (LDH) and complete blood count (CBC) for lymphoma.[11]

Treatment for thymoma includes surgical resection, with the goal of achieving negative margins. Pathologic analysis and subsequent staging are used to determine the need for adjuvant therapies, such as chemotherapy or radiation, as well as to determine the appropriate surveillance plan post-operatively.[12-15] Resection is often performed through an open surgical approach via median sternotomy, though minimally invasive approaches, such as video-assisted thoracoscopic surgery (VATS), have been adopted for smaller masses—usually <5 cm.[16-21] While minimally invasive surgery (MIS) techniques have been used for giant thymoma up to 15cm in the adult literature, there are no case reports detailing a VATS approach for giant thymoma in the pediatric population.[19-21] We present the case of a pediatric, giant thymoma resected via a VATS approach. Close collaboration between adult thoracic surgery and pediatric surgery was vital in the success of this technique.

CASE PRESENTATION

A 17-year-old healthy female presented to pediatric surgery clinic with 2 months of isolated, exertional chest pain. Three maternal family members, across two generations, had a history of thymoma resection via sternotomy. Physical exam was remarkable for reproducible chest pain and electrocardiogram showed possible incomplete right bundle branch block. Labs were remarkable for elevated calcium (10.7 mg/dL) and LDH (278 U/L). AFP and beta-HCG were within normal limits. Chest radiograph was notable for a markedly enlarged cardiac silhouette, particularly in the right chest **(Figure 1)**. Chest CT with contrast revealed a bilobed, anterior mediastinal heterogeneous soft tissue mass with multicystic lesions (left lobe 16.2 x 10.9 x 7.3 cm, right lobe 11.4 x 8.1 x 6.4 cm) and possible communication of the cystic lesions at the level of the

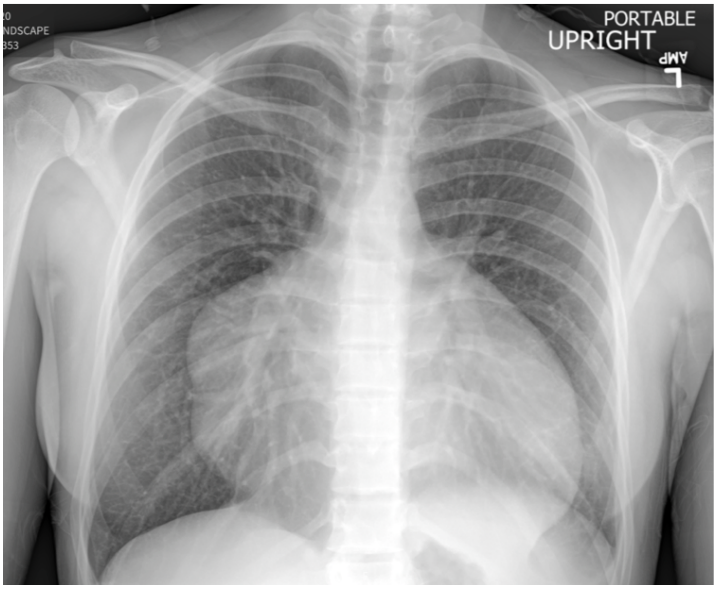

Figure 1. Chest X-ray (CXR) at presentation showing enlarged cardiac silhouette.

pulmonary trunk **(Figure 2)**. These findings, in conjunction with family history, were concerning for thymoma, however the differential included germ-cell tumor, teratoma, and lymphoma. Transthoracic fine needle aspiration revealed mixed lymphoid and epithelioid cells without high-grade cytomorphologic features, compatible with thymoma. A whole body (vertex to toes) Positron Emission Tomography (PET) scan with fluorodeoxyglucose (FDG) radiotracer was performed with max SUV: 6.8 in the primary mass, with no signs of metastasis.

A multidisciplinary approach was taken involving pediatric general surgery, adult thoracic surgery, and the pediatric anesthesia teams. A pre-operative transthoracic echocardiogram showed significant

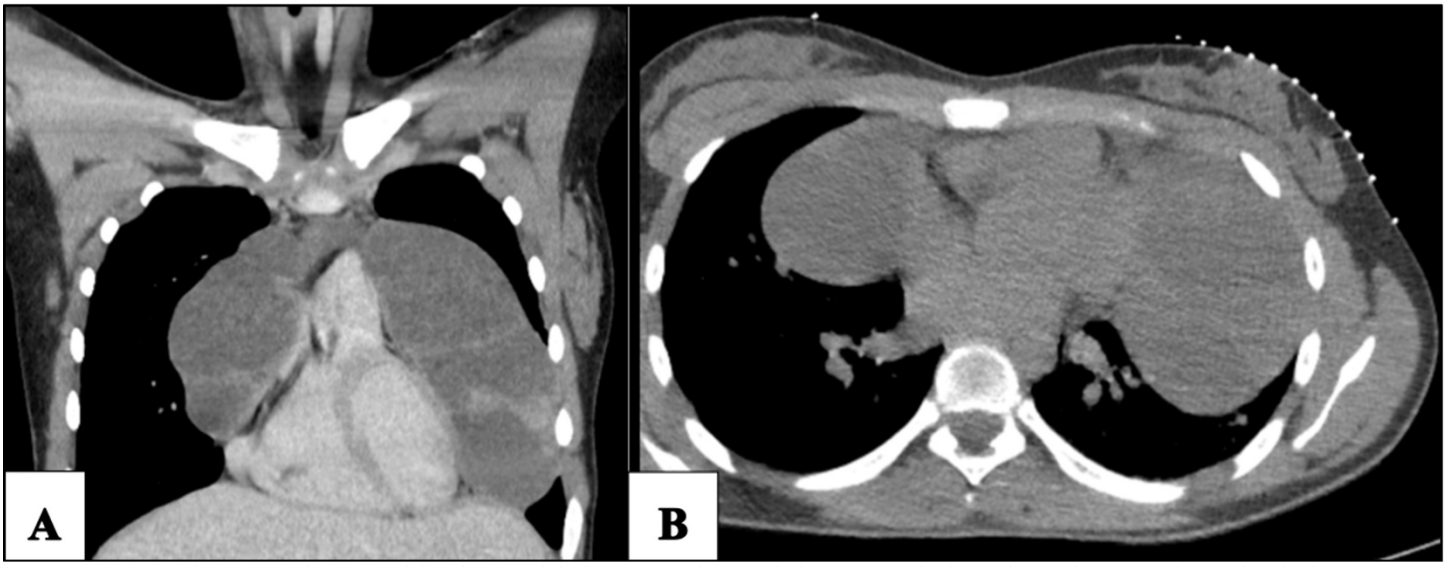

Figure 2. Computed Tomography (CT) of the chest. Axial view (A), and coronal view (B). Both views show a large anterior mediastinal mass crossing midline at the pulmonary trunk.

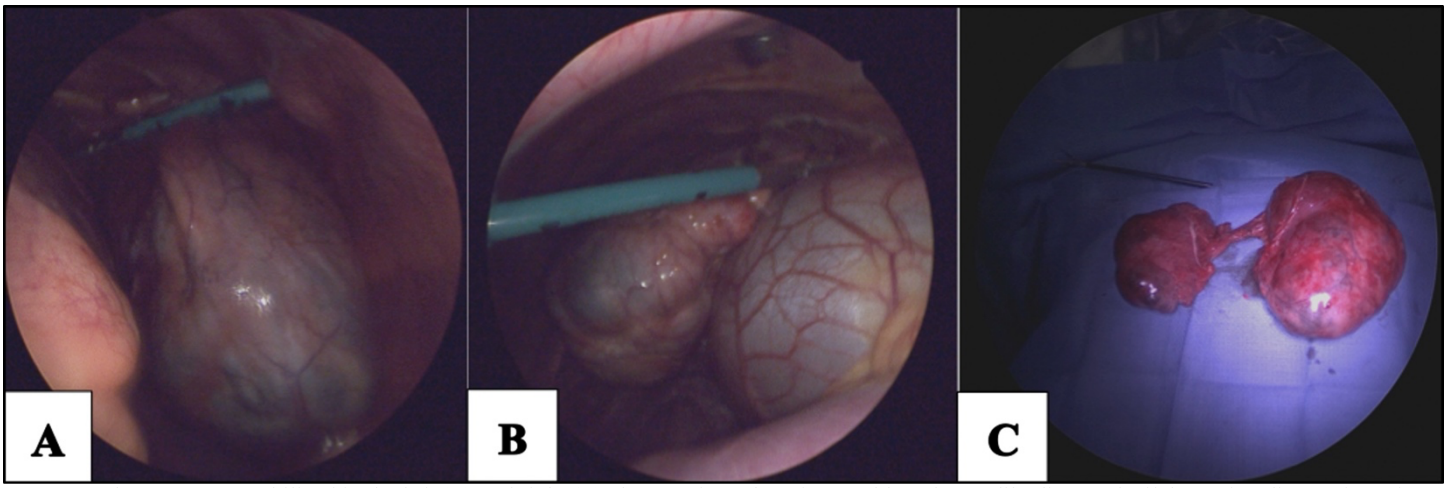

Figure 3. The mass was mobilized in the left (A) and right (B) chest cavity, where it was able to be readily dissected off the pericardium. Dissection planes connected through the mediastinum. Superior mobilization was completed and the mass extracted en bloc through a mini left thoracotomy (C).

compression of the innominate and superior vena cava with increased flow into the right atrium, possible compression of the right atrium by the mass, and dilation of the inferior vena cava, without evidence of any thrombus or obstruction. This guided perioperative resuscitation but did not necessitate any additional intervention. One month after presentation, she underwent resection via a bilateral video-assisted thoracoscopic surgical (VATS) approach with extraction via left mini-thoracotomy. The mass was mobile, with clear planes of dissection off the mediastinal structures and pericardium, with exception of the most superior aspect of the mass. The phrenic nerves were visualized and preserved bilaterally. A muscle sparing left anterolateral thoracotomy (mini-thoracotomy) was performed for final mobilization and extraction **(Figure 3)**. 16 Fr chest tubes were placed bilaterally along with a thoracic epidural for pain control. The left and right lobes measured 16.0 x 10.0 x 7.5cm and 10.5 x 8.0 x 5.0cm, respectively. Pathology review was consistent with well-differentiated thymoma with negative margins. This corresponded to type B3 by World Health Organization (WHO) staging and stage IIb by Masaoka-Koga staging.[22] Immunohistochemistry showed positivity for CK19 and p63.

Her postoperative course was notable for excellent pain control. Initially, a multimodal pain regimen was utilized via bupivacaine epidural, and non-opioid adjuncts. Right and left chest tubes were removed post-operative days (POD) 1 and 2, respectively. Pain scores ranged from 1 to 6; no opioid pain medication was required until the epidural was removed on POD 2. After epidural removal, 5 doses of 5 mg oxycodone were administered in total. She was discharged home on POD 3 in excellent condition. At 6 months, she remains disease-free without need for adjuvant therapy. Surveillance consists of Chest MRI every 6 months for 2 years, then annually for 10 years.

Approximately 12 months after resection, MRI surveillance noted a nodule in the neck, near the thoracic inlet, without evidence of intrathoracic recurrence. Retrospective review of her pre-operative CT and PET did not reveal any visible extension of the index mass into the neck at that time. After consultation with thymoma experts at other institutions, we theorized that this neck mass was residual thymoma, rather than a secondary malignancy. She underwent an open left neck exploration and mass resection, with findings of a 1.5-2.0 cm mass, consistent with thymoma. There were no complications and recovery was unremarkable. She is now transitioning to adult oncology for further follow up.

DISCUSSION

Historically, an open surgical approach, such sternotomy or thoracotomy, has been used as the standard for resection of thymomas greater than 5 cm in size, in both adult and pediatric literature.[23] An open surgical approach has been thought to provide more working space, enhanced exposure, safer dissection of adhesions to major structures, and improved ability to maintain an intact tumor capsule for negative margins.[24] However, in the past 10-15 years, MIS approaches have become the new standard for resection of thymomas and other mediastinal masses.[25] A recent systemic review and meta-analysis reported superiority of MIS approaches, with decreased intraoperative bleeding, operative time, and postoperative complications.[25] In addition, there was shorter ICU length of stay (LOS) and overall LOS, compared to an open approach.[25] Minimally invasive approaches are also associated with faster return to activity and improved engagement in physical therapy.[26]

Pain control is an important aspect of post-operative care following thoracic surgery; another notable aspect of this case was the pain control strategy. Post-operative pain was optimized by the MIS approach. Open approaches, such as median sternotomy or thoracotomy have been shown to produce stronger and longer lasting pain which is more likely to lead to opiate usage and chronic pain.[27] In addition, median sternotomies are not amenable to thoracic epidural blocks, and instead require less common approaches, such as parasternal intercostal plane or rectus sheath blocks, if regional analgesia is desired.[28] Post-operatively, our patient's pain was managed with epidural analgesia, scheduled acetaminophen, and minimal doses of opioid. This allowed a relatively rapid discharge on POD 3, well ahead of the expected 5–11-day length of stay reported for open resections.[16] Remarkably, she did not require any opioid prescriptions at discharge.

MIS approaches to large mediastinal tumors have been used previously, particularly for benign giant thymolipomas.[29,30] However, in these cases, capsular violation does not produce the same downstream consequences as it might in resection of a thymoma.[31] Due to the malignant nature of thymoma, complete resection without capsular disruption is mandatory, requiring more meticulous exposure and dissection.[23,32,33]

While the need for an oncologic resection may historically favor open approach, MIS approaches have been found to be non-inferior, and possibly superior, when it comes to thymoma resection.[34,35] Sicolo et al. reported no significant difference in oncological survival, both

overall and in disease-free groups when comparing open and MIS approaches to thymectomy.[34] Furthermore, a systematic review and meta-analysis suggested that an MIS approach was superior for R0 resection of thymoma.[35]

While MIS approaches have been adopted for thymoma resection, there has been hesitancy in the case of giant thymomas, due to smaller working space, proximity to vital structures, and risk of capsule disruption. Despite these concerns, VATS resection for giant thymoma has been successfully performed in the adult population.[24] All reported pediatric cases of giant thymoma, however, have been resected via an open surgical approach.[4,7,18,36-44] In this patient, a VATS approach was utilized to achieve an R0 resection of a giant pediatric thymoma measuring 16.0 × 10.0 × 7.5 cm and 10.5 × 8.0 × 5.0 cm. Our team considered whether this was the correct approach after discovering residual disease in the neck. An open approach would have consisted of sternotomy; microscopic neck extension would have similarly been left behind. In addition, there was no indication based on pre-operative imaging to approach the mass via a secondary neck incision at that time. In consideration of this, and the benefits of MIS approaches on patient comfort, mobility, aesthetic outcomes, and length of stay, we conclude that a VATS approach was the optimal choice for this patient. This case highlights the safety and efficacy of minimally invasive surgery (MIS) for pediatric mediastinal masses, even of large size. The collaboration of the pediatric surgery and adult thoracic surgery teams pre-operatively and intra-operatively was critical to the success of this surgical approach.

CONCLUSION

Giant thymomas are rare, with an open surgical approach being the historical preference, especially within the pediatric population. This case demonstrates the feasibility of a minimally invasive approach for bilateral, giant thymoma. VATS for a thymoma of this size presents a surgical challenge, requiring multidisciplinary collaboration for successful resection. Pursuit of an MIS approach, paired with an appropriate perioperative pain regimen, allowed for ideal pain management, rapid discharge, and maximal aesthetic outcome, all while achieving optimal oncologic control. While the decision of surgical approach remains patient and surgeon specific, this case encourages surgeons to consider an MIS approach, regardless of tumor size.

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Abbreviations: AFP, Alpha-Fetoprotein, b-HCG, beta-Human Chorionic Gonadotropin, CBC, Complete Blood Count, CT, Computed Tomography, MRI, Magnetic Resonance Imaging, LDH, Lactate Dehydrogenase, MIS, Minimally Invasive Surgery, VATS, Video-Assisted Thoracoscopic Surgery.

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